DOI: 10.4274/gulhane.galenos.2023.49403
Gulhane Med J 2023;65:176-178

Colonic polyposis with underlying diffuse large B-cell lymphoma presenting with ileocolic intussusception

Bryan Zhi Yong Yeoh[1], David Eng Yeow Gan[2], Sentilnathan Subramaniam[2], Nornazirah Azizan[3], Firdaus Hayati[4]

[1]*Department of Surgery, Faculty of Medicine, University of Malaya, Kuala Lumpur, Malaysia*

[2]*Department of Surgery, Queen Elizabeth Hospital, Ministry of Health Malaysia, Kota Kinabalu, Sabah, Malaysia*

[3]*Department of Pathology and Microbiology, Faculty of Medicine and Health Sciences, Universiti Malaysia Sabah, Kota Kinabalu, Sabah, Malaysia*

[4]*Department of Surgery, Faculty of Medicine and Health Sciences, Universiti Malaysia Sabah, Sabah, Malaysia*

Date submitted:
22.11.2022
Date accepted:
19.06.2023
Online publication date:
15.12.2023

Corresponding Author:
Firdaus Hayati, M.D., Assoc. Prof., Department of Surgery, Faculty of Medicine and Health Sciences, Universiti Malaysia Sabah, Kota Kinabalu, Sabah, Malaysia
+60129541429
m_firdaus@ums.edu.my

ORCID:
orcid.org/0000-0002-3757-9744

Keywords: Colon, intussusception, lymphoma

ABSTRACT

Extranodal lymphoma rarely presents as a primary tumor in the gastrointestinal tract, particularly in the form of intussusception in adults. We report the case of a 30-year-old woman admitted with intermittent right lower abdominal pain for one month and a palpable mass on physical examination. Computed tomography revealed an ileocolic intussusception with rectal polypoid mass lesions. The patient underwent a right hemicolectomy because the lead point of the intussusception was identified as a terminal ileum polyp. Histopathological examination revealed diffuse large B-cell lymphoma. In the event of an abdominal mass with radiological evidence of colonic intussusception, surgery remains crucial to avoid unnecessary complications.

Introduction

Extranodal lymphoma is most common in the gastrointestinal tract, mainly in the stomach, followed by the small and large bowel. However, primary lymphoma arising in the gastrointestinal tract is rare. An uncommon sequelae of gastrointestinal lymphoma is intussusception, a condition in which a segment of the bowel telescopes into an adjacent distal segment. This condition mainly occurs in children, while adults comprise less than 6% of cases (1,2). They commonly occur because of intramural or intraluminal lesions that serve as the lead point of intussusception. Here we report an uncommon ileocolic intussusception in a woman with diffuse large B-cell lymphoma (DLBCL) and bowel involvement.

Case Presentation

A 30-year-old female was diagnosed with DLBCL through a biopsy of a nasopharyngeal lesion while being investigated for enlarged cervical lymph nodes. She was pregnant in her third trimester and defaulted chemotherapy after her first cycle because of concerns related to pregnancy complications.

In the fourth postpartum month, she was admitted to the hospital with intermittent right lower quadrant pain for one month. She reported no vomiting, abdominal distension, or blood-stained stool. Bowel-opening frequency and pattern before admission were normal. Abdominal examination revealed a right iliac fossa mass without peritonism. Biochemical tests were normal, with a hemoglobin level of 11.5 g/dL. Screening for human immunodeficiency virus was negative. Computed tomography (CT) revealed an ileocolic intussusception with rectal polypoid lesions (Figure 1A, 1B). Multiple enlarged mesenteric and para-aortic nodes were noticeable. There were no liver or lung lesions. Colonoscopy revealed multiple polyps with mucosal ulcerations in the large bowel (Figure 1C). Biopsy samples were taken from the appropriate lesions.

The patient subsequently underwent exploratory laparotomy, which confirmed the presence of ileocolic intussusception identified by CT (Figure 1D). Multiple polyps were palpable along the large and small bowels with enlarged, hard mesenteric lymph nodes. No stricture or stenosis was observed. A right hemicolectomy (Figure 1E) with ileocolic side-to-side stapled anastomosis was performed. The lead point of intussusception was identified as a terminal ileal polyp. Histopathological assessment of the resected specimen and biopsied polyps were similar to that the nasopharyngeal biopsy, revealing DLBCL, non-germinal subtype (Figure 2A-2D). A total of 12 of 16 lymph nodes were positive for tumor infiltration. The tumor cells were diffusely positive for CD20, BCL6 and MUM1 but negative for CD3, CD10 and BCL2 (Figure 2E, 2F). The Ki67 index was 70-80%. The patient recovered well and was discharged on postoperative day 4 with plans to resume chemotherapy. The patient is well with no recurrence of intussusception until a 6-month clinic follow-up.

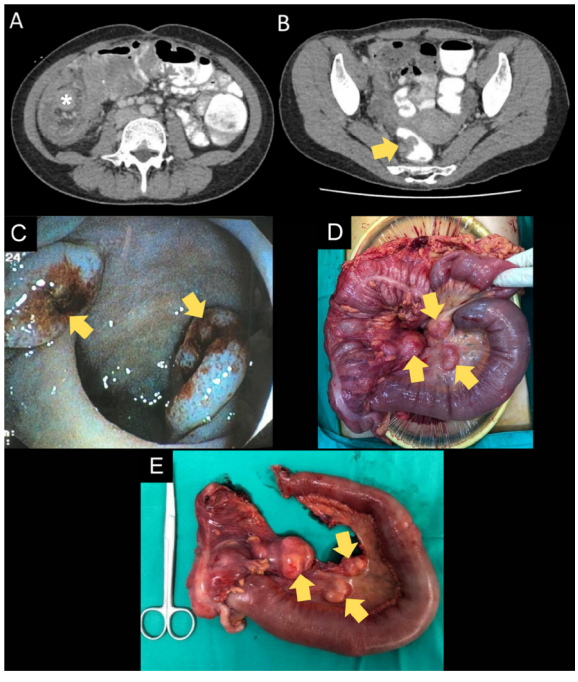

Figure 1. Computed tomography scan of the abdomen showing an ileocolic intussusception (white asterisks) (A) with rectal polypoid lesions (yellow arrow) (B). (C) A colonoscopic finding of multiple rectal polyps (yellow arrows) with mucosal ulceration. (D) Surgery finding of ileocecal intussusception with multiple enlarged mesenteric lymph nodes (yellow arrows). No stricture or stenosis was observed. Polypoidal nodules were palpable throughout the large and small bowel. (E) Post-limited right hemicolectomy specimen showing ileocecal intussusception with multiple enlarged mesenteric lymph nodes (yellow arrows)

Discussion

Intussusception is uncommon in adults. The clinical signs tend to be non-specific, including recurrent abdominal pain, nausea, vomiting, and weight loss. Abdominal pain is present in 95% and tends to be recurrent, frequently lasting longer than 1 week. Acute obstruction or blood-stained stool is less common. A palpable abdominal mass is also infrequent and reported in less than 15% of cases (3). As in our case, clinical course was stormy because her initial chemotherapy plan for DLBCL was affected by pregnancy. The patient then presented with intermittent right lower abdominal pain with a palpable mass during the postpartum period.

The anatomic location of intussusception can be enteroenteric, ileo-ileocolic, or colo-colic. Ileo-ileocolic

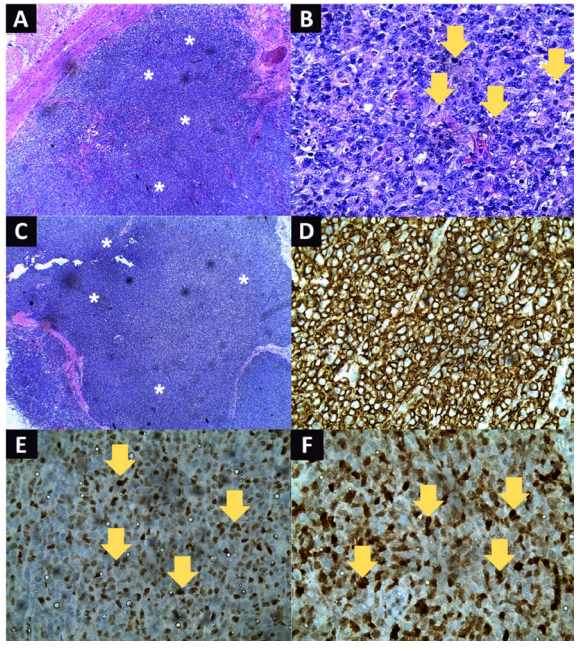

Figure 2. Lamina propria, submucosa, and subserosa (A) infiltrated by sheets of tumor cells (white asterisks) (hematoxylin and eosin, x4 magnification). The tumor cells are medium in size and show hyperchromatic nuclei, coarse chromatin pattern and irregular nuclear contour with mitosis (yellow arrows) (B) (hematoxylin and eosin, x40 magnification). A lymph node infiltrated by tumor cells of similar morphology (white asterisks) (C). Tumor cells are positive for CD20 (D), BCL 6 (yellow arrows) (E), and MUM-1 (yellow arrows) (F). The cells were negative for CD3, CD10, and BCL2 (immunohistochemistry, 40 magnification)

intussusception is the most common site because of its high density of lymphoid tissue (4-6). Endoscopic findings of lymphomatous polyps are variable. They appear as pseudopolypoid lesions, subcentimeter sessile polyps, larger broad-based polyps with ulcerated surfaces, and diffuse infiltration (7). Approximately 10% of cases have gastrointestinal polyposis (8,9). Our patient is interesting because colonoscopy showed synchronous polyps with mucosal ulcerations in the large bowel. The appearance of lymphomatous polyps under narrow-band imaging has not been well studied.

Because up to 63% of adult intussusception cases are related to malignancy (10), resecting the intussuscepted bowel is the mainstay of treatment. However, in the case of transient type, especially in the small bowel, intussusception can be conservatively managed when abdominal symptoms or complications are not found (11). Attempts to reduce the bowel may cause bowel perforation and subsequent tumor seeding in the peritoneum. Furthermore, endoscopic reduction of the intussusception puts the patient at risk of recurrence because the potentially malignant etiology is not excised (12). To the best of our knowledge, the available literature lacks information about recurrent intussusception following bowel resection in patients with multiple polyps. There is also no evidence to support the prophylactic resection of bowel polyps in such patients.

The standard treatment for DLBCL is a combination of cyclophosphamide, doxorubicin, vincristine, and prednisolone (CHOP regime) or with rituximab (R-CHOP) (13). In patients with disseminated gastrointestinal DLBCL (Lugano stage 4), R-CHOP has been shown to improve 3-year overall survival (59%) compared with CHOP (29%), although the difference was not statistically significant (14). No benefit can be obtained in localized disease (14). In disseminated disease, the rate of complete response (CR) to chemotherapy is 46-52%, but the risk of relapse and progression remains high at 40-42%. However, in localized disease, surgery and chemotherapy result in a higher CR rate (85.3% vs. 64.4%, $p<0.001$) (13,14). For any patient who undergoes surgery for gastrointestinal lymphoma, to obtain a promising CR, adjuvant chemotherapy is mandatory. In this study, the patient opted for the CHOP regime during the postpartum period.

Conclusion

Adult intussusception is uncommon and frequently a sequelae of underlying intraluminal or intramural pathology. Even though the standard treatment for DLBCL is chemotherapy, in the event of an abdominal mass with radiological evidence of colonic intussusception, surgical resection remains crucial to avoid unnecessary complications.

Ethics

Informed Consent: The subject provided written consent, and patient anonymity was preserved.

Authorship Contributions

Surgical and Medical Practices: B.Z.Y.Y., D.E.Y.G., S.S., Concept: S.S., Design: N.A., F.H., Literature Search: D.E.Y.G., Writing: B.Z.Y.Y., F.H.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Akbulut S. Unusual cause of adult intussusception: diffuse large B-cell non-Hodgkin's lymphoma: a case report and review. Eur Rev Med Pharmacol Sci. 2012;16:1938-1946.
2. Bernardi D, Asti E, Bonavina L. Adult ileocolic intussusception caused by Burkitt lymphoma. BMJ Case Rep. 2016;2016:bcr2016218334.
3. Kulendran K, Choy KT, Keogh C, Ratnapala D. An Exceptional Case of Ileocolic Intussusception Secondary to Burkitt's Lymphoma: What Variations Are There in the Presentation and Management of Those Patients Who Approach Adolescence? Case Rep Surg. 2018;2018:6251321.
4. Stohlner V, Chatzizacharias NA, Parthasarathy M, Groot-Wassink T. Jejunojejunal Intussusception as the Initial Presentation of Non-Hodgkin's B-Cell Lymphoma in an Adult Patient: A Case Report and Review of the Literature. Case Rep Surg. 2013;2013:619031.
5. Johan S, Lee PP, Azizan N, Hayati F, Zakaria AD. Vanek's tumor causing ileoileal intussusception in a middle-aged man. Oxf Med Case Reports. 2020;2020:omaa050.
6. Tan JS, Teah KM, Hoe VC, et al. Jejunojejunal intussusception secondary to inflammatory fibroid polyp: A rare cause of small bowel obstruction. Ann Med Surg (Lond). 2020;59:251-253.
7. Johan S, Khairuddin A, Zuki AM, et al. Malignant ulcer: a great mimicker of gastric plasmablastic lymphoma. Clin J Gastroenterol. 2021;14:1027-1030.
8. Ghimire P, Wu GY, Zhu L. Primary gastrointestinal lymphoma. World J Gastroenterol. 2011;17:697-707.
9. Martins C, Teixeira C, Gamito É, Oliveira AP. Mantle cell lymphoma presenting as multiple lymphomatous polyposis of the gastrointestinal tract. Rev Bras Hematol Hemoter. 2017;39:73-76.
10. Nam S, Kang J, Park H, Lee KY, Sohn SK. Adult ileocecal intussusception caused by malignant lymphoma. Korean J Clin Oncol. 2014;10:46-48.
11. Mulita F, Tchabashvili L, Liolis E, Maroulis I. Transient small bowel intussusception in a 30-year-old male. Prz Gastroenterol. 2021;16:252-253.
12. Fernandes C, Pinho R, Ribeiro I, et al. Recurrent cecocolic intussusception in a young woman. Gastroenterol Hepatol. 2015;38:586-588.
13. Hwang HS, Yoon DH, Suh C, Park CS, Huh J. Intestinal diffuse large B-cell lymphoma: an evaluation of different staging systems. J Korean Med Sci. 2014;29:53-60.
14. Kim SJ, Kang HJ, Kim JS, et al. Comparison of treatment strategies for patients with intestinal diffuse large B-cell lymphoma: surgical resection followed by chemotherapy versus chemotherapy alone. Blood. 2011;117:1958-1965.